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Case report

Sudden death and laryngeal papillomatosis: A case report

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ABSTRACT

Laryngeal papillomatosis is an uncommon respiratory disease. Often misdiagnosed, it can cause acute respiratory insufficiency, quickly fatal if not promptly faced. A case of sudden death due to laryngeal papillomatosis is described in a woman presenting to the Emergency Department (ED). Laryngeal obstruction was not diagnosed in previous medical examinations nor recognised at the time of attempted intubation in ED; only post-mortem investigation allowed discovering a large fleshy mass occluding the larynx in the region of the vocal folds.

1. Introduction

Acute upper airway obstruction can be secondary to many causes (foreign body, infection, neoplasia, etc.). Circumstantial and anamnestic data, particularly the onset and progression of symptoms, can guide clinicians in differential diagnosis.

In emergency medicine, an important skill is to recognise uncommon diseases that can lead to fatal outcome. An unusual condition, detectable in young children, is juvenile laryngeal papillomatosis; its incidence has been estimated between 3.84 and 7 cases per million per year[1] and its presentation is often an acute airway obstruction, sometimes fatal.[2–5]

Adults can be affected by a less aggressive form; in these cases, the slow progression of the disease increases the possibility of misdiagnosis, more frequently as asthma or allergies.[6]

We report a case of sudden death in a Negroid woman due to asphyxiation caused by a laryngeal papillomatosis not previously diagnosed.

2. Case report

After dinner, a 42-year-old Negroid woman felt a little tired; she decided to take rest in the living room while her husband tidied up the kitchen. Half an hour later, she was found lying on the couch unconsciousness, cyanotic and apnoeic, with her face dirty from regurgitated food. Emergency medical staff, promptly called, tried to free up the airways to provide manual bag-mask ventilation. Regurgitated food persisted at laryngeal aditus; it could not be aspirated because of its particular consistency. The woman arrived at the Emergency Department (ED) in respiratory arrest. Physicians were able to intubate her although with difficulty, but after the placement of the oro-tracheal probe a bloody secretion flowed from the airways. After long-lasting intensive care operations, physicians certified the death. The woman's husband reported that, during the past 6 months, his wife suffered for some episodes of acute onset of respiratory distress attributed to asthma in previous medical examinations.

2.1. Autopsy findings

Post-mortem investigation showed marked narrowing of the larynx, in the region of the vocal folds, due to a large fleshy mass sticking out in the airway lumen (Figs. 1 and 2). Multiple greyish-white, cauliflower-like, friable papillomas completely hid the false and true vocal cords; there were no detectable scars due to surgical trauma or mechanical action. Along the distal airways there were no lesions, but only traces of white thick material similar to the digested food found in the stomach. The neck region was also lacking abnormalities caused by any exogenous traumatic injuries (haemorrhagic infiltrations, laryngeal cartilage damages or bone fractures). Additional autopsy findings included pulmonary oedema, visceral congestion and petechiae on the scalp.

The cause of death was attributed to asphyxiation secondary to obstruction by laryngeal papillomatosis.

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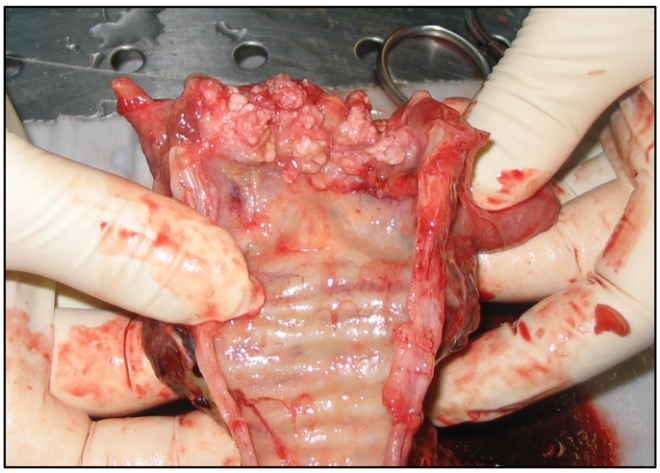

Fig. 1. Autoptic finding of laryngeal papillomatosis: multiple cauliflower-like friable papilloma at vocal folds.

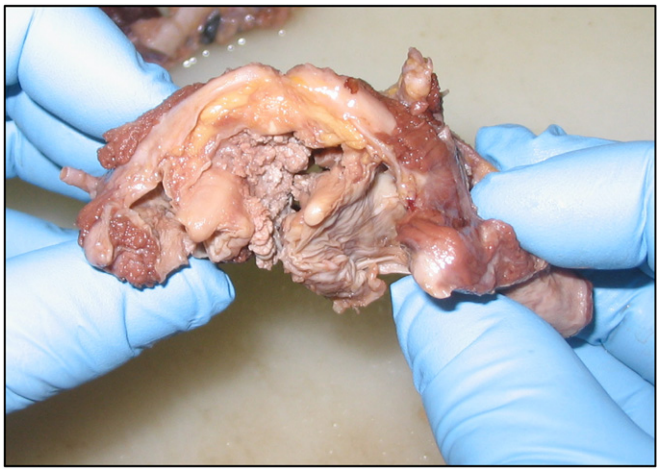

Fig. 2. Formalin-fixed larynx: exophytic proliferation sticking out in the airway lumen.

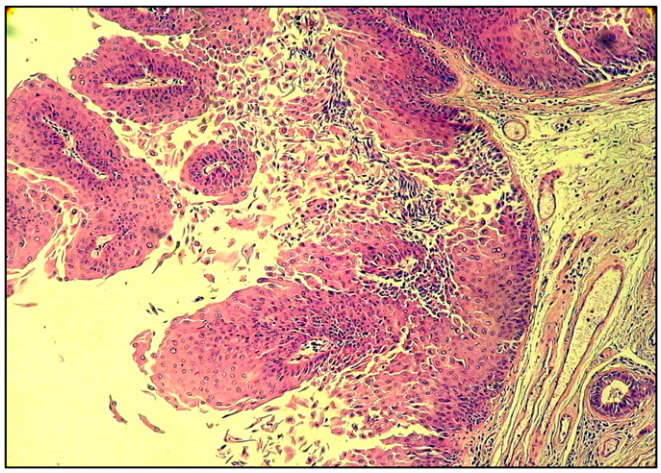

Fig. 3. Histological papillomata architecture: a multilayer epithelium that overlies a fibrovascular core arranged in several branching finger-like projection (EE 100×).

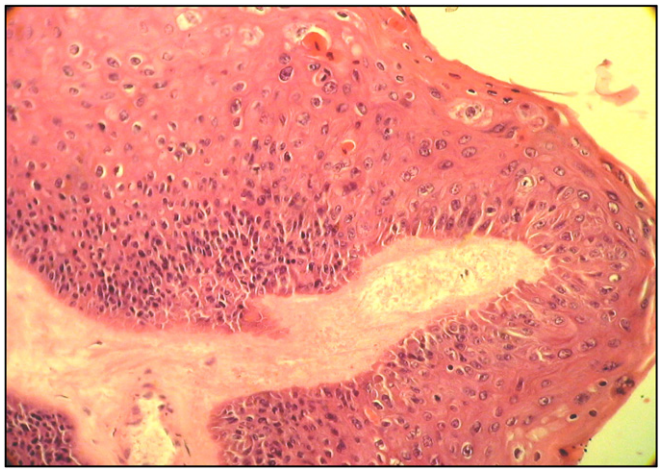

Fig. 4. Multilayer squamous parakeratotic epithelium with koilocytosis (EE 250×).

2.2. Histological findings

Histology confirmed the diagnosis: the laryngeal mass had the typical papillomata architecture with a multilayer, squamous, parakeratotic epithelium that overlaid a fibrovascular core arranged in several branching finger-like projections (Figs. 3 and 4).

The stroma was poorly infiltrated by neutrophils and mast cells. Mast-cell density was assessed immunohistochemically using anti-tryptase antibodies; immunoreactivity demonstrated the absence of degranulated mast cells in the lining epithelium and in connective tissue.

Viral aetiology, as reported in the literature, was supported by the identification of koilocytes, large cells with conspicuous irregularly shaped nuclei and a characteristic perinuclear vacuolisation of the cytoplasm, which suggests the role of the human papillomavirus (HPV) in the cell proliferation process of the laryngeal mass.[7,8]

3. Discussion

The presence of whitish and thick digested food in the mouth and in the upper airways of the woman masked the laryngeal pathology: physicians in ED considered this material as digested food difficult to remove, while it was a laryngeal endoluminal proliferation obstructing the lumen and hindering the placement of the endotracheal probe required for patient ventilation. The woman's ab ingestis prevented a prompt recognition of the laryngeal pathology that could have been easily identified through a direct visualisation by a laryngoscope. If the obstructing lesion was evidenced, a rapid tracheotomy would have been performed after the first intubation attempt.

Asthma anamnesis misled the physicians who mistakenly attributed the woman's acute respiratory failure to it. The literature reports several cases regarding upper airway obstruction misdiagnosed as asthma.[4] An insidious progression of symptoms and an emergency situation (as in this case) cause difficulties in clinical assessment; moreover, the rare occurrence in adults of respiratory papillomatosis makes this diagnosis not so easy.

According to the husband's report, the woman complained of features of sub-acute upper airway obstruction only in the last 6 months. This anamnestic finding agrees with the typical natural history of laryngeal papillomatosis in adults: the progressive growth of the mass determines a clinical silence, broken by the attainment of a critical size that can compromise the patency of the airways.[9] Autoptic investigations confirmed the presence of a sharp reduction of the laryngeal lumen due to the multiple polypoid projections of the laryngeal mass.

The insidious presentation of the disease may also be accelerated by precipitant factors.[10] Inhaled foreign body or alterations in the neck tissues caused by trauma or surgery are able to determine the definitive acute airway obstruction with complete blockage of the respiratory flow. Necroscopic surveys did not reveal the presence of any of them in this case. Upper airway infections or hypersensitivity immune reactions can be precipitant factors too, but histological and immunohistological investigations in this case did not reveal any alterations.

The role played by food regurgitation is uncertain: it is not possible to rule out that ab ingestis determined the final mechanical obstruction of the airways, or that it was only a secondary event due to acute respiratory failure.

When upper airway obstruction occurs, laryngeal papillomatosis, although rare, must be remembered in differential diagnosis.

Ethical approval
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Conflict of interest statement
None.

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